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# REVIEW ARTICLE

# Sarcoma and Metastatic Carcinoma

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Primary tumors of the bone and soft tissue of the pelvis are rare. Proper surgical treatment requires a fundamental knowledge of the biology of malignant musculoskeletal neoplasms. This understanding allows stratification of sarcomas into a staging system. In addition to prognostic value, the careful staging of the neoplasms dictates the type of surgical margins necessary and guides in the use of adjuvant therapy. Limb salvage techniques developed for the reconstruction of major extremity structural deficits can be used for reconstruction of the pelvis. This review first addresses the biologic behavior and staging of malignant musculoskeletal neoplasms. The surgical techniques employed for the resection and the reconstruction of the pelvis are then discussed.
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**KEY WORDS: sarcoma; staging; biopsy; surgery; reconstruction; arthroplasty**

## INTRODUCTION

Primary tumors of bone and soft tissue are rare, accounting for <1% of the new tumors diagnosed each year in the United States [1] . It is estimated that the annual incidence of bone sarcoma is 1 per 100,000 [2,3]. Osteosarcoma, the most common primary malignancy arising in bone (excluding multiple myeloma) presents in the pelvis <10% of the time [2].

In contrast, carcinoma occurs with relatively frequency. Prostate, breast, lung, thyroid, kidney, and bladder carcinomas are associated with a propensity toward skeletal metastases. The pelvis, with its large areas of cancellous bone, is a common site for skeletal metastases.

Generally, surgery of sarcomas arising in the bones and soft tissues of the pelvis is of curative intent, requiring wide or radical margins. Resections are difficult and the reconstructions demanding. A few patients with isolated skeletal metastatic foci of carcinoma as well as a few carefully selected patients with visceral carcinoma directly invading the sacrum, ilium, or ischium may benefit from large resections of curative intent as well. Generally, all other patients with metastatic carcinoma require surgery only for palliation of pain or when there is significant risk for pathologic fracture.



## SARCOMA

Primary neoplasms that occur in the muscles and bones of the pelvis are sarcomas and by definition are of mesenchymal origin. They are named for the matrix they produce. For example, a malignant neoplasm producing cartilage is termed a chondrosarcoma. A malignant neoplasm producing bone is termed an osteosarcoma. Likewise, a malignant tumor arising in the soft tissues with a fibrous matrix is a fibrosarcoma.

Sarcomas behave in a predictable fashion. Their growth is in a centrifugal manner until a potential barrier to spread is encountered. Dense fascial planes, the origins and insertions of muscles and tendons, cortical bone, and articular cartilage serve as barriers and are the basis for anatomic compartments. Unfortunately, in the pelvis these barriers are not as well developed, nor are the compartments as easily defined as they are in the extremity ( i.e., the anterior compartment of the thigh). As the tumor grows, it compresses surrounding host tissue inciting a host response of fibrovascular infiltrate creating a reactive zone. Only truly benign tumors are encapsu-

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lated, while aggressive benign and malignant tumors are surrounded by a pseudocapsule within the reactive zone. Finger-like projections of the primary tumor called "satellite" lesions are found within the reactive zone. Twenty-five percent of high grade tumors are associated with "skip" lesions. Skip lesions are by definition, a microscopic or macroscopic focus of tumor within the compartment of origin that is separated from the primary neoplasm by normal host tissue (lesion is located outside of the reactive zone) [4,5]. Skip lesions represent hematogenous metastases within the compartment of origin.

## STAGING

The Musculoskeletal Tumor Society (MTS) staging system is used for staging primary neoplasms of bone and can be used for the staging of soft-tissue sarcomas [6,7]. Benign lesions are designated numerically as stage 1, 2, or 3. Malignant tumors are designated with Roman numerals as stage I, II, or III. Malignant tumors are further classified as to whether they are contained within their compartment of origin (A: intracompartmental) or have crossed into the adjacent compartment (B: extracompartmental).

Stage 1 benign lesions are latent, inactive. They frequently are asymptomatic and are discovered incidentally. Most benign lesions present as stage 2 active and frequently may have pain or other symptoms associated. Stage 1 and stage 2 lesions are both surrounded by a true capsule. Stage 3 aggressive benign lesions are symptomatic with initial complaints of pain and discomfort. They may have local inflammatory sign and may present with a pathologic fracture. Their capsule is incomplete, and they are surrounded by a pseudocapsule. Frequently, stage 3 lesions cross compartmental boundaries.

The MTS staging system should be considered to be a surgical staging system. It is based not only on the histologic grade, but also on the radiographic appearance and the clinical behavior of the neoplasm. Low-grade malignant neoplasms are designated stage I. Stage I malignant neoplasms may present asymptomatically, but pain is a common complaint. Histologically, although they produce a well-differentiated matrix, they have cytologic markers of malignancy. Stage I malignant neoplasms most frequently present intracompartmentally (stage IA) and are rarely associated with distant metastases. Chondrosarcoma is an example of a sarcoma that frequently presents as a stage IA lesion. High-grade malignant neoplasms are designated stage II. High-grade lesions are usually symptomatic and may present with a pathologic fracture. Histologically they are poorly differentiated and exhibit markers of malignancy such as anaplasia and abnormal mitoses. Stage II malignant tumors quickly extend through compartmental boundaries and are associated with a significant incidence of pulmonary metastases. Conventional osteosarcoma frequently presents as a stage IIB lesion. Stage III is used to designate all lesions that present with metastases. These include pulmonary, distant, regional (lymphatic) and skip lesions.

The staging system is useful in guiding surgical margins necessary for control. There are by definition 4 margins obtainable: (1) Intralesional—the plane of dissection passes through the tumor. Gross disease is left behind. (2) Marginal—The plane of dissection is through the reactive zone. "Satellite" lesions within the reactive zone are may be left behind. (3) Wide—the plane of dissection passes through normal host tissue. The tumor and the entire reactive zone are removed. However, skip lesions are potentially left behind. (4)Radical—the resection removes the entire compartment from origin to insertion. The tumor, the reactive zone, and any associated skip lesions are removed [4,5].

For stage 1 and stage 2 benign lesions, a marginal margin surrounding the encapsulated neoplasm yields a low recurrence rate. Often an adjuvant such as cryotherapy or the thermal effects of polymethylmethacrylate is used in stage 2 bony lesions to "extend" an intralesional margin after a careful curettage. Stage 3 aggressive benign tumors transgress their capsule and therefore are associated with satellite lesions within the reactive zone. They are often treated with a wide margin as one would treat a low-grade malignancy. Rarely, adjuvant external beam radiation is used in the pelvis when resection would carry significant morbidity.

Stage I (A and B) low-grade sarcomas require a wide margin. Wide resection of pelvic lesions is complicated by the close proximity of major neurovascular structures and the pelvic viscera. Adjuvants such as chemotherapy and external beam radiation are not generally effective for low-grade sarcomas. Stage II high-grade sarcomas require radical margins. When an effective adjuvant exists, a wide margin can be used facilitating limb salvage. All gross disease including the reactive zone is resected widely through normal tissue relying on the adjuvant to control any microscopic skip lesions.

## RADIOGRAPHY

Conventional radiography of the bony pelvis begins with a routine anteroposterior radiograph of the pelvis. Additional views may be taken to add specific information. Inlet and outlet views allow better visualization of the pelvic ring and sacrum, while Judet views add information concerning the iliac wings, the acetabulum, and supporting structures of the acetabulum. Often a great deal of information can be obtained from a tumor's conventional radiographic appearance [8]. Bone destruction and incomplete attempts at containment may be present. Primary bone tumors may produce a matrix that has a

typical radiographic appearance. Stippled or punctate calcifications are seen in cartilaginous neoplasms. The presence of intralesional calcifications when coupled with other radiographic signs such as cortical destruction can be indicative of a chondrosarcoma. Tumoral bone formation can be indicative of an osteogenic sarcoma, particularly when extra-osseous and associated with a soft tissue mass. Several carcinomas are known to be associated with bony metastases, including prostate, breast, lung, thyroid, kidney, and bladder. Carcinoma of the thyroid and kidney typically produce osteolytic metastases, while carcinoma of the prostate is associated with osteoblastic metastases. Carcinoma of the breast may produce osteolytic and osteoblastic metastases [9].

Technetium-99m scintigraphy (bone scan) is very useful both as an initial screening tool when metastatic spread to the pelvis is expected as well as in the evaluation of primary bone lesions. Almost all primary neoplasms arising in bone show increased activity on a bone scan. In addition, the degree of uptake can aid in formulating a differential diagnosis and can indicate occult involvement of the bony pelvis with deep-seated soft tissue sarcomas. Although multiple myeloma is classically thought of as cold on bone scan, it often reveals areas of increased uptake due to surrounding reactive bone formation.

Computed tomography (CT) scanning is useful in defining the integrity of the cortical bone [10]. CT is particularly helpful in the pelvis where the three-dimensional anatomy can make conventional radiography difficult to interpret. CT is helpful in determining the nature of the matrix produced by a lesion. It can also demonstrate if the lesion is well defined and surrounded by reactive bone or if the lesion is poorly defined and exhibits a permeative pattern and aggressive destruction of surrounding bone. Additionally, CT angiography may be of benefit in defining the location and direct involvement of surrounding major vascular structures.

Magnetic resonance imaging (MRI) is useful to define the soft tissue extent of a lesion as well as its intramedullary extent [11–13]. MRI is also useful for determining the presence of skip lesions. Multiple lesions associated with metastatic carcinoma and multiple myeloma are readily demonstrated. Magnetic resonance angiography is an emerging technology that may be of benefit in the preoperative planning of pelvic tumors as well.

Other specialized imaging modalities are occasionally performed when indicated. After the completion of all imaging studies, including screening for metastases, a differential diagnosis is formulated. Based upon the imaging studies, the benign or malignant nature of a lesion and at times the histologic diagnosis can be predicted with reasonable certainty [14].

## BIOPSY

A biopsy is generally necessary to confirm a diagnosis. Too often, the biopsy is performed with little thought and foresight when, in fact, the biopsy requires careful preoperative planning. The biopsy should be performed only after all initial staging studies have been completed and should be performed by the surgeon who will perform the definitive procedure. It is well documented that a patient's ultimate morbidity and oncologic outcome are significantly altered when the biopsy is performed inappropriately [15–19]. The preoperative planning for the biopsy is often is as extensive as the preoperative planning for the definitive resection. With sarcomas, the biopsy tract (including needle tract) must be excised completely with the tumor. In most cases of suspected primary bone neoplasms, an incisional biopsy will need to be performed to ensure adequate material is obtained for permanent sections as well as for special stains, cytogenetics, electron microscopy, etc. Occasionally, a trephine or core biopsy may be performed [17, 20]. A frozen section should be performed at the time of open biopsy to ensure that adequate tissue representative of the neoplasm has been obtained. If a hypervascular lesion such as a metastatic renal cell carcinoma is suspected, preoperative angiography and embolization should be performed as life-threatening hemorrhage can occur even through a relatively small biopsy site. When certain neoplasms are suspected, such as metastatic carcinoma or multiple myeloma, a CT-guided needle biopsy may be sufficient for diagnosis. Soft tissue sarcomas arising in the gluteal or abductor musculature can often be diagnosed by Tru-cut (Travenol Laboratories, Inc., Deerfield, IL) needle biopsy under local anesthesia. Minimally invasive biopsy techniques should be performed with the same care and planning as an open biopsy.

## SURGERY

Hemipelvectomy was once considered the only option available for sarcomas arising within the bones and soft tissues of the pelvis [21,22]. Although this disfiguring operation provides a satisfactory outcome from an oncologic standpoint, it is coupled with significant functional impairments. Often pelvic tumors reach such extremely large size that it is difficult to obtain radical or wide margins even when a hemipelvectomy is performed [23,24]. While the tumor locally provides much difficulty, the limb distal to the tumor is usually normal, making it difficult to justify its amputation if it can be spared. Applying the principles of limb salvage surgery learned in the extremities, it has become apparent that the resection of pelvic neoplasms (internal hemipelvectomy) can be performed with a resultant margin and oncologic outcome equal to that of a conventional or extended

hemipelvectomy [3,25–27]. Most series have reported an acceptable local recurrence rate when an appropriate margin has been obtained [19,25,28–31]. Limb salvage surgery of the pelvis carries a higher complication rated than that of the extremity, but the rate of complications is more dependent upon the reconstruction chosen rather than the resection itself [32]. Even with the impairment associated with pelvic reconstruction after internal hemipelvectomy, the patient's level of function is significantly better than if an amputation had been performed [3,33]. Limb salvage surgery of the pelvis (internal hemipelvectomy) is now accepted and is generally applied to primary bone and soft tissue tumors arising in the pelvis [24,34–36]. Carefully selected patients with visceral tumors that invade the bony pelvis may now benefit from resection and salvage of the extremity as well [37–41].

## RESECTION

Limb salvage surgery involves three distinct phases—resection, skeletal reconstruction, and soft tissue reconstruction [42,43]. The resection is performed based upon the margin needed for tumor control. The skeletal reconstruction is chosen based upon the defect remaining as well as the patient's oncologic prognosis coupled with his overall physical state, age, physical demands, and willingness to accept a higher complication rate to perhaps obtain a higher level of function. Recently, the trend may be shifting from more complex reconstructions toward less complex reconstructions attempting to decrease the complication rate. Careful attention is given to soft tissue reconstruction of the pelvic floor and inguinal region to decrease the incidence of postoperative hernia and to fill potential dead spaces to decrease the incidence of post-operative hematoma with secondary infection. Occasionally, local muscle flaps must be used to provide satisfactory coverage.

Resections of the pelvis are classified by the method of Enneking and Dunham [31]. A type I resection removes the supra-acetabular ilium to the sacroiliac joint. A type IA resection adds the abductors of the hip and the sciatic nerve if necessary. A type II resection removes the acetabular region from the sciatic notch to the lateralmost portion of the pubic rami. A type IIA resection adds an extra-articular resection of the hip joint including the proximal end of the femur. A type III resection involves all or part of the pubis with a type IIIA adding the femoral neurovascular bundle.

## SURGICAL APPROACHES

Perhaps the most useful approach is a utilitarian incision described by Enneking and Shirley [31]. Prior to surgery, the bladder is catheterized and if significant posterior dissection is anticipated, the bowel is prepared. The incision starts over the posterior superior iliac spine and then extends anteriad along the iliac crest to the point of the femoral neurovascular bundle. The incision then extends distad along the rectus femoris for several inches and then curves gently posteriad to end lateral to the shaft of the femur at its junction of proximal and middle third. This allows the development of a large posterior myocutaneous flap that may be used to close a conventional hemipelvectomy if the need were to arise immediately or later. Care is taken to spare the superior gluteal artery if possible.

The entire external and internal hemipelvis with the exception of the pubic rami can be exposed. If further medial exposure is needed, a limb can be created along the inguinal ligament to the midline or to the contralateral side if needed (in essence incorporating an ilioinguinal or Pfannenstiel approach) [35,44,45]. The inguinal canal is unroofed. In the female, the round ligament is divided, and in the male, the spermatic cord is protected, retracting it toward the midline. The floor of the inguinal canal is then incised just proximal to the inguinal ligament and the deep dissection carried to the symphysis pubis where the rectus femoris is released. The external iliac vessels are exposed to the common iliac vessels or to the aorta and inferior vena cava if necessary.

The ureter is identified and protected. The femoral nerve is identified where associated with the iliopsoas muscle and is protected. The retropubic space of Retzius is entered with careful retraction of the bladder. If dissection proceeds to the midline anteriorly, care is taken to identify the urethra beneath the symphysis pubis. The iliacus is taken as additional margin on the medial surface of the ilium. Although the iliacus may be "thinned" with a large soft tissue component of a pelvic sarcoma arising within the bone, its fascial covering usually provides a satisfactory margin. If exposure medially is not needed past the pectineal eminence, the descending limb of the incision can be moved laterally to the anterior superior iliac spine, and the deep dissection extended along the plane between the reflected head of the rectus femoris and the vastis lateralis as in an extended iliofemoral approach [46].

For large tumors of the retroperitoneum with pelvic side wall involvement, an abdominoinguinal incision may be used. A midline abdominal incision is connected to a vertical incision crossing the inguinal ligament and extending distally over the femoral triangle. By dividing the insertion of the rectus abdominus and incising the floor of the inguinal canal, complete dissection of the iliac vessels may be performed [47]. In all cases, an adequate margin must be obtained, and therefore formal orthopaedic approaches utilizing internervous planes are rarely used in there entirety [48]. Figure 1 demonstrates convenient sites of pelvic osteotomy.

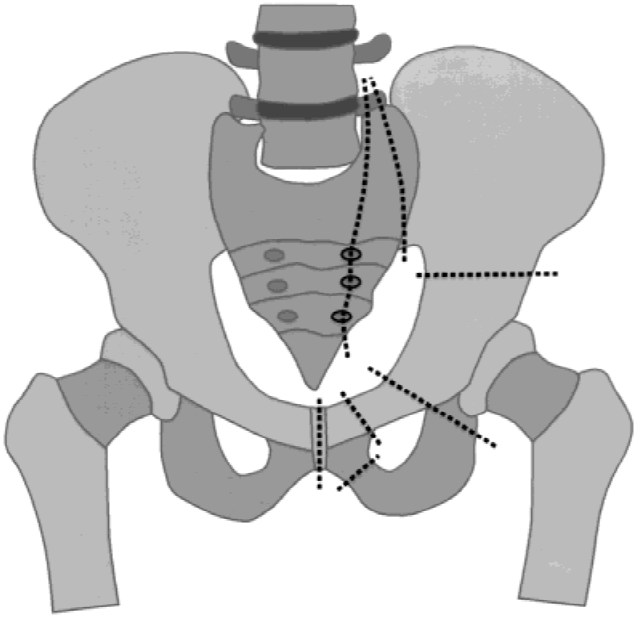

Fig. 1. Convenient osteotomy sites.

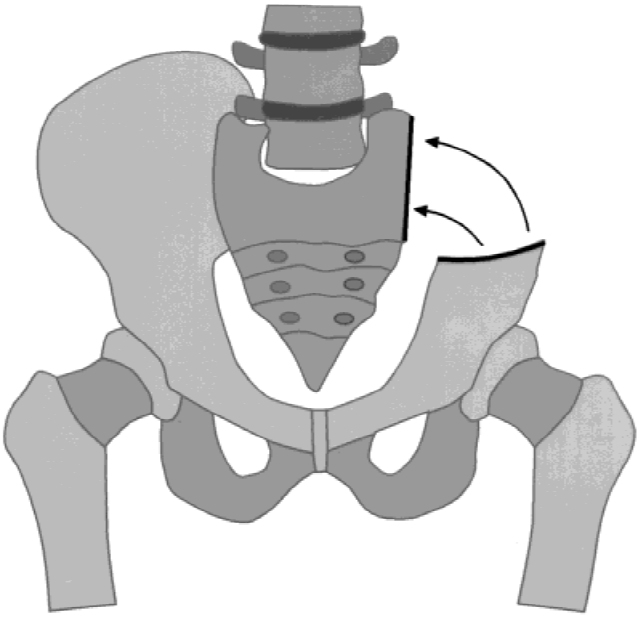

Fig. 2. Reconstruction of type I pelvic resection.

## RECONSTRUCTION

The available options for skeletal reconstruction of the pelvis largely depend upon the type of resection. When there has been no disassociation of the pelvic ring, reconstruction often is unnecessary. When an intralesional margin has purposely been obtained as in the case of some benign lesions and in most metastatic carcinomas, the options available to fill the defect are autograft bone, allograft bone, commercially available bone void fillers, and polymethylmethacrylate (PMMA). Bone graft does not incorporate well in the face of active tumor growth. Therefore polymethylmethacrylate is generally used in the case of metastatic disease in which the procedure is generally palliative. Additionally, PMMA has the benefit of immediate structural stability allowing immediate weight bearing. PMMA is mostly used in the periacetabular region when the patient is thought to be at significant risk of pathologic fracture. Long Steinman pins can be driven into the more proximal uninvolved ilium and sacral ala to aid in the support of a acetabular component of a total hip arthroplasty. A protrusio cup is placed against the pins and cemented into place filling the bony defect [49–51]. It is prudent to expect metastatic disease to progress. Therefore, when planning a reconstruction, one should anticipated loosening and failure of standard components, and techniques that provide longer term stability should be employed such as above.

Surgical resection of sarcoma requires wide or radical margins. A few carcinomas may benefit from wide resection of isolated skeletal metastases. Because of the magnitude of bone and soft tissue removed, these reconstructions are the most challenging to the surgeon.

### Type I Resection

Type I resections may allow the simplest reconstruction. Soft-tissue coverage is not a problem unless a type IA resection has been performed. Following a type I resection, the remaining portion of the anterior ring of the pelvis can be rotated to close the gap to the sacrum. The symphysis pubis acts as a hinge and limited internal fixation is used to obtain fusion (Fig. 2). A small leg length discrepancy is produced. After a type IA resection, the iliopsoas is transferred to the greater trochanter creating an abduction moment [31].

### Type II Resection

Type II resections involve resection of the acetabulum, and this is technically the most challenging. Options include a flail extremity, pseudarthrosis, arthrodesis, composite allograft total hip arthroplasty, custom design total hip arthroplasty, saddle prosthesis, reimplantation of autoclaved bone with arthroplasty, and rotationplasty.

Function of a flail extremity is poor, and although some ambulatory ability can be regained, this is generally reserved for a nonambulator or as a palliative procedure [52]. An arthrodesis can be performed, its location dependent upon the remaining bone stock [19,24,31]. An iliofemoral arthrodesis may be easier to obtain than an ischiofemoral arthrodesis, but results in significant leg length discrepancy (Fig. 3). When a solid iliofemoral arthrodesis is obtained, the sacroiliac joint allow some hip flexion as there are no restraining anterior structures. Although hip flexion is still very limited, it allows easier sitting than a conventional arthrodesis of the hip in which the pelvic ring is intact. Although an ischiofe-

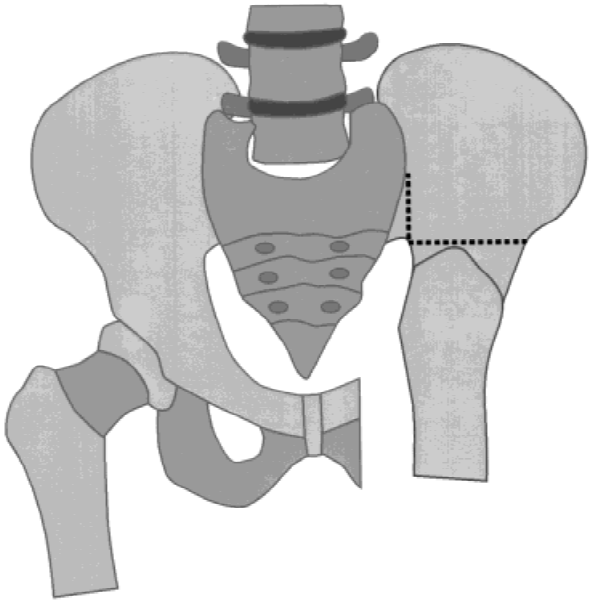

Fig. 3. Reconstruction of type II pelvic resection with iliofemoral arthrodesis.

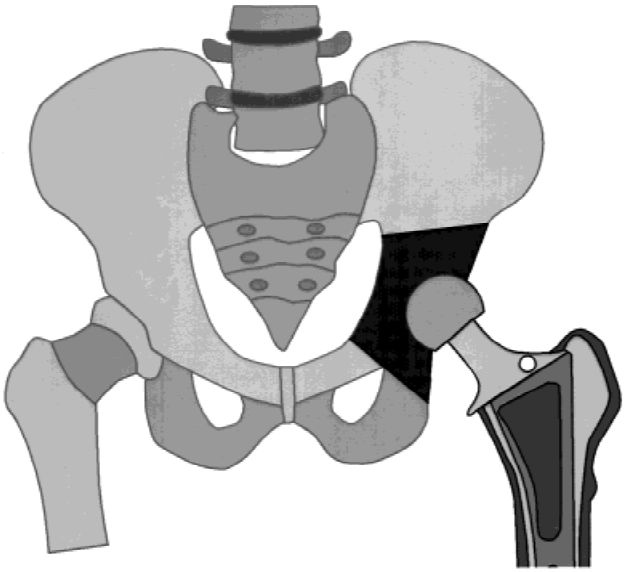

Fig. 4. Reconstruction of type II pelvic resection with allograft prosthetic composite reconstruction.

moral arthrodesis may provide a better gait because of more equal leg lengths, some patients experience significant pain at the symphysis pubis. The most common complication of arthrodesis is failure to obtain union [19].

If a type IIA resection has been performed including a significant portion of the proximal femur, an intercallary allograft is required to complete the arthrodesis.

Pseudarthrosis can be achieved similarly to an arthrodesis. Again the location of the pseudarthrosis is dependent upon the remaining bone stock. The patient is secured with limited fixation, generally in the form of malleable wire or cable and is protected post-operatively for some duration of time. A stable pseudarthrosis may allow more movement at the hip while possibly decreasing pain when performed anteriorly at the pubis.

The use of an allograft or reimplantation of autoclaved resected bone reconstitutes the integrity of the pelvic ring [27,53]. The use of autoclaved bone is appealing in that there is no size mismatch inherent with massive pelvic allografts. The bone is fixed to pelvis using standard internal fixation techniques and a cemented total hip arthroplasty is performed (Fig. 4). Complications inherent to the autoclaved bone include stress fractures and deep infection. The amount of resected soft tissue may leave a conventional unconstrained total hip arthroplasty unstable with a significant dislocation rate, while a constrained total hip arthroplasty may lead to a higher rate of loosening. An allograft is fixed in similar fashion [54,55]. The allograft restores continuity of the pelvic ring and maintains leg length as well as provides a functional hip joint through the use of conventional total hip arthroplasty techniques. It unfortunately has met with a high rate of infection and failure [30,32,56–58].

Various types of custom total joints have been employed. These range from those that reconstitute the pelvic ring incorporating an acetabular component to those that attach to the remaining ilium thus restoring axial-femoral continuity [23,59–61]. Computer-aided design modeling has improved the design and fit. However, difficulty arises if the plane of osteotomy of the pelvis differs from that planned preoperatively. Generally these allow immediate weight bearing but suffer from the usual complications associated with total hip arthroplasty such as loosening.

A variation of arthroplasty is the saddle prosthesis. A saddle-like projection is attached to a convention total hip arthroplasty stem and rests in a notch in the remaining portion of the ilium [62]. The saddle prosthesis was originally developed for large defects surrounding revision total hip arthroplasty, and its experience in that capacity has shown the best function is obtained when the abductors and the iliopsoas are maintained. The saddle prosthesis maintains leg length but only allows limited flexion. Complications center around dislocation and deep infection. The saddle prosthesis allows relatively early weight bearing and is not dependent on bony union, therefore strengthening its appeal in a patient with a limited life expectancy.

Occasionally, sarcoma occurs in the pelvis of the pediatric patient. The use of prosthetic joints and allografts is limited by the amount of future growth anticipated. If significant growth remains, a rotationplasty may be performed [63]. The tumor is resected in bloc with the proxi-

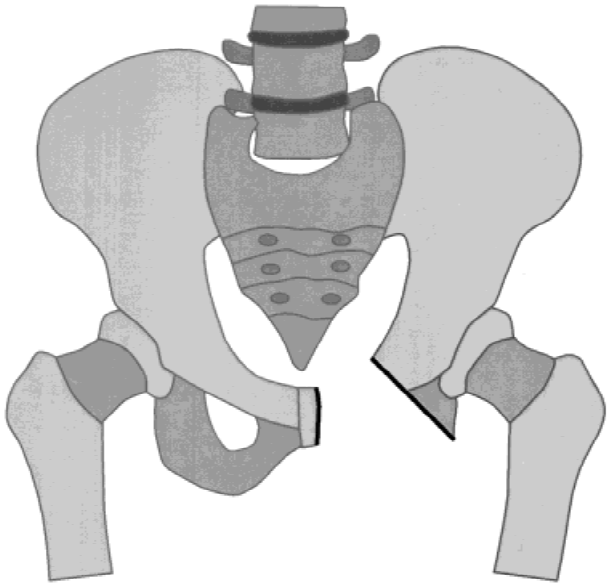

Fig. 5. Type III pelvic resection: no bony reconstruction necessary.

mal femur preserving the vascular supply to the limb and its innervation. The distal femur and limb are rotated 180° and secured to the remaining pelvis. The knee now functions as the hip joint and the ankle as the knee joint. The patient is fitted with a modified below knee amputation prosthesis. A rotationplasty is durable and essentially unrestricted activity is allowed.

### Type III Resection

Generally, reconstruction of the bony pelvis is not required (Fig. 5). However, meticulous detail should be given to the closure of the soft tissues of the pelvic floor with consideration for local muscle flaps.

## CONCLUSION

A sarcoma arising in the pelvis represents one of the most difficult challenges facing the surgeon. Using principles based upon the biologic behavior of sarcomas, it is now possible to resect these tumors with predictable oncologic outcomes. It is no cliché that the treatment of the complex malignancies is multidisciplinary. In addition to the musculoskeletal oncologist and surgical oncologist, the treatment often includes the plastic and reconstructive surgeon, the urologist, the medical oncologist, and/or the radiation oncologist. Because of the complexity involved in the treatment of these rare malignancies and the significant risk of negatively influencing functional outcome and survivorship, only tertiary institutions that frequently deal with sarcomas should embark on the care of these patients. Additionally, any surgical care other than palliative for metastatic carcinoma to the pelvis or visceral carcinoma with direct pelvic extension should likewise be referred for care at a tertiary facility.

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